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Main Article

Treatment of idiopathic laryngotracheal stenosis with laryngotracheal reconstruction

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Abstract

Objectives: We evaluated the efficacy of laryngotracheal reconstruction with costal cartilage grafting for the treatment of idiopathic laryngotracheal stenosis.

Methods: From January 2001 to December 2005, 129 patients with laryngotracheal stenosis were treated at our hospital. Of these patients, five (4 per cent) female patients whose ages ranged from 14 to 34 years were identified as having idiopathic laryngotracheal stenosis. These patients were treated with a modified laryngotracheal reconstruction with anterior costal cartilage grafting. A Montgomery T-tube remained in place for 12 to 22 months.

Results: Three patients presented with grade three and two with grade two stenosis. Three patients were decannulated after one procedure, with normal respiratory function and good exercise tolerance; one patient was decannulated after two procedures. One patient failed decannulation. The mean time to decannulation was 13 months.

Conclusions: Laryngotracheal reconstruction with anterior costal cartilage grafting is a safe and effective method, and provides an alternative treatment for idiopathic laryngotracheal stenosis.

Key words: Larynx; Trachea; Stenosis; Reconstructive Surgical Procedures

Introduction

Laryngotracheal stenosis still remains a therapeutic challenge. In the adult population, this condition is commonly caused by mechanical trauma from endotracheal intubation, external trauma, upper respiratory tract infection, systemic disorders (e.g. Wegener's granulomatosis) and neoplasms.

However, in a small number of cases the cause of subglottic stenosis cannot be determined. These patients usually have a long history of progressive shortness of breath, hoarseness and wheezing. The usual macroscopic appearance of the lesion is circumferential, fibrotic stenosis that is limited to the cricoid cartilage and the first one or two tracheal rings, usually occurring in female patients.[1,2] This condition is termed idiopathic laryngotracheal stenosis.

Many different treatment methods have been used for idiopathic laryngotracheal stenosis, ranging from conservative therapies to definitive surgical procedures.[3–6] Conservative treatment involves local corticosteroid injections, airway dilatation and laser ablation. Definitive surgical procedures include laryngotracheal segmental resection and primary anastomosis, which have been considered to have good to excellent long-term results. However, the optimal management strategy in the treatment of idiopathic laryngotracheal stenosis remains controversial. Treatment with laryngotracheal reconstruction has been reported in a few cases.[1]

In this paper, we present our experience in treating five cases of idiopathic laryngotracheal stenosis using laryngotracheal reconstruction, over a five-year period.

Materials and methods

From January 2001 to December 2005, 129 patients with laryngotracheal stenosis were treated at our hospital. Of these patients, five (4 per cent) female patients were identified as having idiopathic laryngotracheal stenosis. Their mean age was 23 years (range, 14 to 34 years). The time interval from onset of symptoms to diagnosis ranged from 10 to 36 months (mean, 21.8 months). These patients' follow-up times ranged from 24 to 76 months (mean, 54.6 months).

The diagnosis of idiopathic laryngotracheal stenosis was made by excluding known causes of subglottic stenosis. All patients were tested for antineutrophil cytoplasmic antibodies to rule out Wegener's granulomatosis. No patients had gastroesophageal reflux disease. No patient gave a history of intubation for general anaesthesia in the two years before onset of

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symptoms. Flexible fibre-optic laryngoscopy and biopsy were performed to exclude neoplasm, granuloma, infection, vasculitis and other systemic connective tissue disorders as causes. Plain X-rays and computed tomography scanning of the larynx and upper trachea were performed to assess the extent and severity of the lesion. Stenosis was graded according to the Myer-Cotton grading system.[7]

Previous treatment had included laser ablation in one patient, laryngeal enlargement procedures with T-tubes in two patients and the same without T-tubes in one patient. Four patients were tracheostomy-dependent. These treatments had been performed at other institutions.

Laryngotracheal reconstruction with costal cartilage grafting was undertaken. This operation has been well described by Cotton and Zalzal and Cotton.[8,9] Briefly, a mid-line incision was made anteriorly in the cricoid and upper trachea. A boat-shaped costal cartilage graft was used to expand the airway, placed anteriorly with stenting. A Montgomery T-tube was placed, and remained in situ for 12 to 22 months. We modified the operation by removing any scar tissue and repairing the defect with a free skin graft.

Results and analysis

A summary of patient data is shown in Table I. Of the five patients, three presented with grade three and two with grade two stenosis. The stenosis was almost always located in the region between the upper edge of the cricoid and the lower edge of the second tracheal ring.

Four patients were able to be decannulated, with normal respiratory function and good exercise tolerance. One patient failed decannulation. Three patients were decannulated after one procedure and one after two procedures (the failed patient underwent two procedures). The mean time to decannulation was 14.8 months (range, 12 to 22 months).

In the one patient not extubated (patient one), severe scar tissue was present from the subglottis to the second tracheal ring and almost no cricoid or tracheal rings could be identified, because of previous procedures.

For patient three, we used only laryngotracheal reconstruction with scar tissue removal and free skin grafting, instead of costal cartilage grafting, because of the mild degree of the lesion.

Patient two was extubated nine months after surgery but failed decannulation. She required a second laryngotracheal reconstruction procedure, and a T-tube remained in place for a further 13 months, before final decannulation.

Patients three, four and five required only one procedure, and were decannulated after 12 months of stenting.

The most common complication was granulation tissue formation in the trachea and epiglottis. Four patients had granulation tissue growth requiring laser removal (one to three applications).

Discussion

The aetiology of idiopathic laryngotracheal stenosis is still unclear.

TABLE I

PATIENT DATA: SUMMARY

Pt	Age (y)	Sex	Grade*	Prev therapy	Trache before referral?	Symptom duration (mths)	FU (mths)	Surgery	Stent duration (mths)	Outcome	Post-T-tube therapy
1	17	F	III	Cricoid split + scar tissue resection + no stent × 2	Y	27	74	LTR + CCG + T-tube	16	Failed	Laser† removal of granulations
2	19	F	III	Cricoid split + scar tissue resection + T-tube stent × 2	Y	10	61	LTR + CCG + T-tube stent × 2	22‡	Decannulation	Laser† removal of granulations × 3
3	34	F	II	None	N	36	76	LTR + T-tube stent	12	Favourable	None
4	32	F	II	Laryngofissure with scar tissue resection + T-tube stent	Y	25	38	LTR + CCG + T-tube stent	12	Decannulation	Laser† removal of granulations
5	14	F	III	Laser + dilation + no stent × 2	Y	11	24	LTR + CCG + T-tube stent	12	Decannulation	Laser† removal of granulations × 2

*Myer-Cotton classification. †Nd:YAG. ‡Nine months then 13 months. Pt = patient; y = years; prev = previous; trache = tracheostomy; mths = months; FU = follow up; F = female; Y = yes; LTR + CCG = laryngotracheal reconstruction with anterior costal cartilage grafting

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Oestrogen is considered to be the major cause, as almost all patients reported in the literature have been female. However, Benjamin *et al.* and Dedo and Catten found no oestrogen receptors in samples taken from the lesion area in patients with idiopathic laryngotracheal stenosis.[1,3]

Another possible cause is long-term gastroesophageal reflux disease. Jindal *et al.* reported six patients with idiopathic laryngotracheal stenosis who had stabilised and responded to surgical management after evaluation for gastroesophageal reflux disease followed by medical management.[10] However, other authors speculated that if gastroesophageal reflux disease were the underlying cause, one would expect many more male patients, and also recurrence of stenosis in the absence of reflux treatment.[3,11] In Grillo's series of 73 patients, none developed recurrent stenosis over many years, despite receiving no treatment for gastroesophageal reflux disease. None of our patients had symptoms or signs related to gastroesophageal reflux disease.

Idiopathic laryngotracheal stenosis is a diagnosis of exclusion. Biopsy and histopathological examination are important to exclude known causes such as tuberculosis, histoplasmosis and amyloid disease. Antinuclear cytoplasmic antigen testing is helpful to exclude Wegener's granulomatosis. When all known causes have been excluded, a diagnosis of idiopathic laryngotracheal stenosis can be made. As the signs and symptoms are mild at the onset of the disease, it usually takes a long time to make a final diagnosis. In our series, a mean time of 21.8 months was observed between onset of symptoms and final diagnosis.

There is currently no consensus on the optimal treatment of idiopathic laryngotracheal stenosis.

Benjamin *et al.* treated 12 patients with endoscopic laser vaporisation, and were successful in maintaining the airway of eight patients with an average of 4.2 laser procedures per patient (range, two to eight).[1]

Dedo and Catten used operative techniques (endoscopic laser submucosal resection and mucosal flap rotation) in 50 patients.[13] Most patients required continued treatment, with an average number of six procedures each, while 13 patients failed to decannulate.

Some authors consider endoscopic laser-assisted dilatation and scar tissue resection to be the treatments of choice for initial management.[1,4,12]

In repeated failures, open neck surgery with laryngoplasty or laryngotracheal resection and anastomosis is recommended.

Lee and Rutter recently reported a series of six patients treated by balloon dilatation.[13] The airway was dilated to 2.0 to 3.5 endotracheal tube diameters larger than its initial width. Four patients were followed for 10 to 30 months, without recurrence.

Grillo and colleagues have advocated definitive treatment with single-stage laryngotracheal resection.[11] In their series of 73 patients, all were successfully decannulated; 67 (91 per cent) had good to excellent long-term results in terms of voice and breathing quality, and did not require further intervention.

We used laryngotracheal reconstruction with anterior costal cartilage grafting to treat idiopathic laryngotracheal stenosis. This procedure is a popular and effective method of widening the airway lumen in paediatric cases of laryngotracheal stenosis, but it has not been confirmed as useful in the management of adult, idiopathic laryngotracheal stenosis.[1,12]

Both Cotton and Rethi have emphasised that scar tissue should not be removed during the procedure, since to do so would produce a raw intraluminal surface.[8,14] Idiopathic laryngotracheal stenosis has a unique histopathological appearance, with bland inflammatory fibrosis of the keloidal type, a thickened lamina propria and intact cartilage.[5] Therefore, we chose to resect the scar, in order both to widen the airway lumen and to remove the pathological tissue; we then repaired the defect by free skin grafting.

It is our experience that the Montgomery T-tube should remain in place for a relatively long time, although there is no optimal duration of stenting following laryngotracheal reconstruction. Stern *et al.* reported a duration of stenting ranging from two weeks to 23 months in paediatric patients, with a mean of 7.4 months.[15] In patients with idiopathic laryngotracheal stenosis, a long period of stenting may be needed in order to hold the cartilage and skin grafts in position and to prevent scar contracture. In our series, all patients wore a T-tube for at least 12 months. Patient two failed to decannulate after nine months of stenting, and required a second laryngotracheal reconstruction procedure and stenting for a further 13 months before ultimate decannulation.

- **Idiopathic laryngotracheal stenosis has been treated by various surgical techniques; effective management by laryngotracheal reconstruction with anterior costal cartilage grafting has not been confirmed**
- **Laryngotracheal reconstruction was found to be a relatively safe and effective method, and thus provides an alternative treatment for idiopathic laryngotracheal stenosis**
- **Removal of scar tissue and repair of the defect with a free skin graft, and a long period of T-tube stenting, may be important for achieving decannulation**

For patients with severe scar tissue formation, laryngotracheal reconstruction with cartilage grafting may not be suitable. Our patient one was such a case, failing due to previous, unsuccessful procedures and severe scar formation in the subglottic and upper tracheal regions. This patient eventually refused to receive any further therapy, including further laryngotracheal resection.

Conclusion

Laryngotracheal reconstruction with anterior costal cartilage grafting is a relatively safe and effective method that provides an alternative treatment for

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idiopathic laryngotracheal stenosis. Removal of scar tissue and repair of the defect by free skin graft, plus a long period of T-tube stenting, may be important for achieving decannulation. Given the small number of patients in this series, a larger study would be needed to further support these conclusions.

References

1 Benjamin B, Jacobson I, Eckstein R. Idiopathic subglottic stenosis: diagnosis and endoscopic laser treatment. *Ann Otol Rhinol Laryngol* 1997;**106**:770–4
2 Ashiku SK, Mathisen DJ. Idiopathic laryngotracheal stenosis. *Chest Surg Clin North Am* 2003;**13**:257–69
3 Dedo HH, Catten MD. Idiopathic progressive subglottic stenosis: findings and treatment in 52 patients. *Ann Otol Rhinol Laryngol* 2001;**110**:305–11
4 Valdez TA, Shapshay SM. Idiopathic subglottic stenosis revisited. *Ann Otol Rhinol Laryngol* 2002;**111**:690–5
5 Grillo HC, Mark EJ, Mathisen DJ, Wain JC. Idiopathic laryngotracheal stenosis and its management. *Ann Thorac Surg* 1993;**56**:80–7
6 Ashiku SK, Kuzucu A, Grillo HC, Wright CD, Wain JC, Lo B *et al*. Idiopathic laryngotracheal stenosis: effective definitive treatment with laryngotracheal resection. *J Thorac Cardiovasc Surg* 2004;**127**:99–107
7 Myer CM 3rd, O'Connor DM, Cotton RT. Proposed grading system for subglottic stenosis based on endotracheal tube sizes. *Ann Otol Rhinol Laryngol* 1994;**103**:319–23
8 Cotton R. Management of subglottic stenosis in infancy and childhood. Review of a consecutive series of cases managed by surgical reconstruction. *Ann Otol Rhinol Laryngol* 1978;**87**:649–57
9 Zalzal GH, Cotton R. A new way of carving cartilage grafts to avoid prolapse into the tracheal lumen when used in subglottic reconstruction. *Laryngoscope* 1986;**96**:1039
10 Jindal JR, Milbrath MM, Shaker R, Hogan WJ, Toohill RJ. Gastroesophageal reflux disease as a likely cause of "idiopathic" subglottic stenosis. *Ann Otol Rhinol Laryngol* 1994;**103**:186–91
11 Grillo HC, Mathisen DJ, Ashiku SK, Wright CD, Wain JC. Successful treatment of idiopathic laryngotracheal stenosis by resection and primary anastomosis. *Ann Otol Rhinol Laryngol* 2003;**112**:798–800
12 Giudice M, Piazza C, Foccoli P, Toninelli C, Cavaliere S, Peretti G. Idiopathic subglottic stenosis: management by endoscopic and open-neck surgery in a series of 30 patients. *Eur Arch Otorhinolaryngol* 2003;**260**:235–8
13 Lee KH, Rutter MJ. Role of balloon dilation in the management of adult idiopathic subglottic stenosis. *Ann Otol Rhinol Laryngol* 2008;**117**:81–4
14 Rethi A. An operation for cicatricial stenosis of the larynx. *J Laryngol Otol* 1956;**70**:283–93
15 Stern Y, Willging JP, Cotton RT. Use of Montgomery T-tube in laryngotracheal reconstruction in children: is it safe? *Ann Otol Rhinol Laryngol* 1998;**107**:1006–9

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